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ORIGINAL ARTICLE *Quality of life*

# A pilot study on the effects of the transition of paediatric to adult health care in patients with haemophilia and in their parents: patient and parent worries, parental illness-related distress and health-related Quality of Life

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**Summary.** The aim of this pilot study was to investigate the effects of the transition from paediatric to adult health care services in haemophilia patients and their parents. We compared pretransition children ( $n = 9$ ) and their parents ( $n = 18$ ) to posttransition patients ( $n = 8$ ) and their parents ( $n = 21$ ). Pre- and posttransition patients did not differ in self-rated health-related quality of life (QoL) or worries about the transition. Fathers of posttransition patients rated their son's QoL as poorer than those of pretransition patients ( $P = 0.034$ ) and

indicated higher levels of illness-related distress than fathers of pretransition patients ( $P = 0.034$ ). The findings indicate that the transition affects parents more than patients. Moreover, we found gender differences in parental worries about the transition. The findings indicate that programmes designed to facilitate the transition in haemophilic patients should also address the patients' parents.

**Keywords:** haemophilia, psychosocial effects, quality of life, transition

## Introduction

Haemophilia is a severe X-chromosome linked coagulation disorder that is characterized by spontaneous bleeding, particularly in muscles and joints, and bleeding in the case of an injury or trauma [1,2]. The illness occurs in mild, moderate and severe forms. The latter is associated with strong impairments in the patients' daily life. To date, there is no cure and therapy focuses on 'on demand' or prophylactic supplementation of the lacking coagulation factor in order to prevent lasting joint damage and resulting disability [1,2]. The treatment is invasive, not only because of the frequent infusions in the case of prophylactic therapy, but also because of the high demands on the patient and his environment [2]. As in other severe chronic illness (e.g. [3,4]), due to

general improvement in medical science and practice (in western countries), the number of children that survive haemophilia through adolescence has increased during the second half of the 20th century (e.g. [5]). These children 'graduate' from the paediatric department around the age of 18 and receive further treatment from adult health facilities.

The transition from paediatric services to adult health care is a serious risk factor for deterioration of the medical condition and (health-related) quality of life (QoL, e.g. [6,7]). From a theoretical point of view, characteristics of both paediatric and adult health care services, and parent and patient characteristics each can account for specific obstacles that can hamper a successful transition (e.g. [6,8–13]). The risk of non-compliance increases in the transition-phase from paediatric services to adult health care (e.g. [6]). Given the invasive and demanding character of the treatment, this may be of particular relevance for patients with haemophilia because a joint bleeding caused by non-compliance may have life-long consequences for the patients. Therefore, one can argue that children with haemophilia and their parents would benefit from empowerment

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programmes that facilitate this transition. Programmes have been developed by different paediatric disciplines to facilitate the transition to adult health care. However, little patient-based empirical data exist to underscore these programmes [11,14,15]. Moreover, empirical data on the efficacy of these programmes are almost absent [16].

This study was designed to explore the impact of the transition from paediatric to adult health care facilities in patients with haemophilia and their parents. Findings from the study may help in successfully innovating the transition programme. We investigated the effects of the transition on the QoL as reported by patients with haemophilia and by their parents. We compared self-reported QoL and worries about the transition of pretransition patients to those of posttransition patients. In addition, we assessed the parents' estimations of their son's QoL, parental worries about the transition and parental illness-related distress.

Assuming that the transition from paediatric care to adult health care can negatively affect the patients' QoL (e.g. [6]), we expected that pretransition patients and their parents report better QoL compared to posttransition patients and their parents. In addition, we expected that pretransition overall worries and illness-related distress scores would be lower than posttransition scores. Furthermore, we investigated to what extent worries and parents' illness-related distress about the transition influence health-related QoL.

## Materials and methods

### *Participants*

Patients and their parents were recruited via the paediatric haemophilia care centres of the University Medical Center Groningen, the University Medical Center St Radboud Nijmegen, the Leiden University Medical Center, the Academic Medical Center Amsterdam, the VU Medical Center Amsterdam and the Catharina Hospital Eindhoven. With the exception of two clinics, all major medical centres in the Netherlands that treat patients with haemophilia participated in the study. Of the two hospitals that did not participate, one hospital recently started a large survey among all its patients in the transition phase; the other is a categorical hospital where paediatric and adult patients with haemophilia receive treatment at the same patient ward (i.e. there is no transition from paediatric to adult health care services). Inclusion criteria were a diagnosis of moderate or severe haemophilia [factor VIII or IX

(FVIII or FIX), <5%] and patients' age older than 14 years. Exclusion criteria were somatic co-morbidity, established psychiatric co-morbidity, inability to complete questionnaires (e.g. because of language difficulties), patients' use of psycho-active medication, and the transition completed more than 5 years ago. Finally, to avoid possible effects of earlier (indirect) experiences of patients and of parents with the transition to adult health care services we also excluded those patients with an elderly brother with haemophilia.

### *Procedure*

Participants were recruited between May 2006 and May 2007. The patients' medical therapist briefly informed the patients and their parents about the study and asked for permission for the researcher to contact the patients and their parents. The researcher, who provided detailed information about the study and asked for their participation, contacted patients and parents who gave permission. After consent had been obtained, the patient and his parents each received a (prenumbered) booklet with the questionnaires and prepaid return envelope.

### *Instruments*

*Worries about the transition.* Dutch translation (first author) of the John Hopkins Adult Cystic Fibrosis Program Survey (7-item parent version and 6-item patient version [17]): assesses the participants' worries about: leaving the physician, meeting new caregivers, receiving poorer care in the adult unit, adult caregivers being less caring, being exposed to infections and being admitted at an adult facility. An additional item was added for parents (their son being responsible for his medical condition by himself). These items may be a point of concern not only patients and their parents prior to transition but also in those after transition (see also [17]). Items are scored on a 5-point Likert scale (ranges from not at all to very). A mean worry score was computed (sum of the individual items/number of items, range 1–5). In addition, a mean interpersonal worries score (mean score of leaving physician, meeting new caregivers and adult caregivers being less caring, range 1–5) and a mean medical worries score (mean score of receiving poorer care, being exposed to infections, admission at an adult facility, and patient being responsible for medical condition, range 1–5) were computed.

*Health-related quality of life.* Dutch version (first author) of the Haemo-QoL-A, versions for adoles-

cents, adults (18+) and for their parents (© 1903 MEDTAP® International and Bayer Biological Products [18]). The Haemo-QoL-A consists of 41 items that assess 6 dimensions of health-related quality of life in patients with haemophilia (physical functioning, role functioning, worry, consequences, positive affect and treatment concerns, all range 0–100%). In addition, a total QoL score was computed (range 0–100%).

*Parents' illness-related distress (van Dongen-Melman [19]).* The questionnaire consists of 52 items that assess 4 domains of illness-related distress in parents: related uncertainties and insecurity, loss of control with respect to the sick child, loss of control with respect to other children and illness-related fears (all dimensions range 0–100%). A mean score can be computed from the scores on these domains that reflects the parents' illness-related distress (range 0–100%, [19]). The original version (designed for parents of children with cancer) was adapted for purpose of use in parents with children with haemophilia.

#### *Statistical analyses*

The data were analyzed by the use of non-parametric statistics (Mann–Whitney *U*-test, and Wilcoxon matched pairs signed ranks test). In case significant effects were observed on the total QoL and the parents' illness-related distress score, we investigated which dimensions contributed to this effect. The data were tested 2-sided with  $\alpha$  set at 0.05.

## **Results**

### *Subjects*

Fifteen pretransition families and 14 posttransition families agreed to participate in the study. Of these, nine children and their parents prior to transition (mean age patients:  $15.3 \pm 1.1$  SD; mother:  $46.4 \pm 3.7$  SD; father:  $46.6 \pm 3.5$  SD) and eight patients and their parents who had completed the transition (mean age patients:  $19.4 \pm 2.1$  SD; mother:  $48.5 \pm 4.8$  SD; father:  $50.3 \pm 4.6$  SD) completed the questionnaires. In addition, we received completed questionnaires of three mothers (age 47, 49 and 52) and two fathers (age 53 and 54) of patients who had completed the transition. Their sons failed to complete the questionnaires. For posttransition patients, the mean number of months since transition was 27.8 ( $\pm 19.0$  SD). In two cases, the parents were divorced. In one case, the child's biological father participated,

in another case the child's stepfather participated. The participants under study are 64% of the original sample of patients and parents who initially agreed to participate. One family reconsidered their agreement to participate. In the other cases lack of time was the main reason for drop out.

### *Patient and parent worries about the transition*

Table 1a presents the mean worries of pretransition patients and of their parents with respect to the transition. In contrast to our expectations, neither pre- and posttransition patients nor the parents of pre- and posttransition patients differed in reported worries about the transition (Mann–Whitney *U*-tests). Patients' and fathers' worries about changes in the interpersonal contacts with the physician did not differ from their worries about changes in the quality of medical treatment (Wilcoxon Matched Pairs Signed Ranks Test) but mothers' medical worries exceeded their interpersonal worries about the transition (Wilcoxon matched pairs signed ranks test:  $Z = -2.03$ ,  $P = 0.043$ ). Also, mothers tended to be more worried about the transition than sons (Wilcoxon matched pairs signed ranks test:  $Z = -1.94$ ,  $P = 0.052$ ) and fathers (Wilcoxon matched pairs signed ranks test:  $Z = -1.79$ ,  $P = 0.074$ ). More detailed analyses revealed that mothers were more worried about medical aspects of the transition than sons (Wilcoxon matched pairs signed ranks test:  $Z = -2.71$ ,  $P = 0.007$ ) and fathers (Wilcoxon matched pairs signed ranks test:  $Z = -2.53$ ,  $P = 0.011$ ). A comparison of the patients with haemophilia under study to the patients with cystic fibrosis studied by Boyle *et al.* [17] showed that the latter are more worried about the transition than the haemophilic patients under study (*T*-test; mean patient worries reported by Boyle 2.98; mean worries reported by the patients under study 1.52,  $t = -10.44$ ,  $df = 16$ ,  $P < 0.001$ ).

### *Parental distress*

Table 1b presents the parental illness-related distress scores for mothers and fathers. No differences were observed in mothers in pretransition and posttransition illness-related distress ( $Z = -1.10$ ,  $P = 0.271$ ). However, posttransition fathers showed more illness-related distress than pretransition fathers ( $Z = -2.13$ ,  $P = 0.034$ ). Analyses on the levels of dimensions revealed that posttransition fathers reported higher illness-related uncertainties ( $Z = -2.12$ ,  $P = 0.034$ ) and higher illness-related fears ( $Z = -2.29$ ,  $P = 0.028$ ) than pretransition

**Table 1.** Mean patients worries about the transition and self rated quality of life, mean maternal and paternal worries about the transition, illness-related distress, and estimated quality of life of their sons for all participants and for pre- and post-transition groups separately. For statistics please see text.

	All		Pretransition		Posttransition	
	Mean ( <i>n</i> )	SD	Mean ( <i>n</i> )	SD	Mean ( <i>n</i> )	SD
(a) <i>Worries about the transition</i>						
Patients mean worries	1.52 (17)	0.58	1.50 (9)	0.56	1.54 (8)	0.63
Mothers mean worries	1.88 (20)	0.89	1.79 (9)	0.73	1.95 (11)	1.03
Fathers mean worries	1.65 (19)	0.57	1.52 (9)	0.38	1.76 (10)	0.70
Patient interpersonal worries	1.51 (17)	0.61	1.56 (9)	0.60	1.46 (8)	0.67
Mothers interpersonal worries	1.77 (20)	1.04	1.81 (9)	0.91	1.73 (11)	1.17
Fathers interpersonal worries	1.68 (19)	0.87	1.52 (9)	0.53	1.83 (10)	1.10
Patient Medical worries	1.53 (17)	0.68	1.44 (9)	0.62	1.63 (8)	0.77
Mothers medical worries	2.04 (20)	0.93	1.83 (9)	0.72	2.19 (11)	1.08
Fathers medical worries	1.62 (19)	0.47	1.53 (9)	0.38	1.70 (10)	0.54
(b) <i>Illness-related distress</i>						
Mothers	27.05 (20)	17.27	22.30 (9)	14.44	30.94 (11)	19.04
Fathers	27.80 (19)	17.03	19.86 (9)	17.72	34.94 (10)	13.46
(c) <i>Quality of life</i>						
Patients	86.31 (17)	10.76	89.19 (9)	8.86	83.07 (8)	12.34
Mothers	82.98 (20)	13.46	88.70 (9)	8.26	78.30 (11)	15.37
Fathers	81.04 (19)	13.26	88.44 (9)	9.17	74.38 (10)	13.15

fathers. Paternal distress scores did not differ from maternal distress scores ( $Z = -0.24$ ,  $P = 0.809$ ).

#### *Health-related quality of life*

The QoL scores of patients and their parents are presented in Table 1c. The total QoL score in pre-transition patients did not differ from the total QoL score in posttransition patients ( $Z = -1.19$ ,  $P = 0.232$ ). Hence, our expectation that the transition affects QoL was not supported by our data. Mothers of posttransition patients tended to rate their sons' QoL as lower compared to mothers of pretransition patients ( $Z = -1.79$ ,  $P = 0.074$ ), although the difference was not significant. Fathers of posttransition patients rated their sons' QoL as significantly lower than fathers of pretransition patients ( $Z = -2.12$ ,  $P = 0.034$ ). Analyses of specific dimensions revealed that fathers of posttransition patients rated the physical functioning and treatment concerns of their son as lower than fathers of pretransition patients.

#### *Associations between worries, illness-related distress and quality of life*

Table 2 presents the Spearman rank order correlations between patients' and their parents' worries, parental distress, and patients' and parents' estimated QoL. As shown in this table, the patients' self-rated QoL and the parents' estimations of their child's QoL are interrelated. Furthermore, the patient's self-rated QoL is negatively correlated with patient worries

about the transition. Paternal and maternal estimations of their son's QoL are negatively correlated with their worries about the transition and with both their own and their partner's illness-related distress. The paternal estimation of the son's QoL is also negatively correlated with their son's worries about the transition. For mothers, this association is not significant. Finally, patient and parental worries about the transition and parental distress were interrelated. Fisher  $Z$  tests [20] revealed that the observed associations did not differ between pre- and posttransition groups.

#### **Discussion**

In this paper, we investigated the effects of the transition from paediatric to adult health care facilities in a sample of Netherlands' patients with haemophilia and their parents. In contrast to our expectations, pre- and posttransition patients did not differ in either their worries about the transition or in their self-reported QoL. Pre- and posttransition parents, however, were found to differ. Fathers of posttransition patients reported higher illness-related distress than those of pretransition patients. In addition, they estimated their son's QoL as lower than fathers of pretransition patients. Posttransition mothers also tended to estimate their son's QoL as poorer than mothers of pretransition patients. Finally, we found that mothers tended to be more worried about the transition than both patients and fathers, specifically with respect to the medical issues around the transition.

**Table 2.** Spearman rank order correlations between patients' self-reported QoL and parents' estimations of their child's QoL, patients' and parents' worries about the transition, and parents' illness-related distress. Only significant correlations are presented.

	Patient QoL	Maternal QoL	Paternal QoL	Patient worries	Maternal worries	Paternal worries	Maternal distress	Paternal distress
Patient QoL	*							
Maternal QoL	$R = 0.60, P = 0.012$							
Paternal QoL	$R = 0.75, P = 0.001$	*						
Patient worries	$R = -0.56, P = 0.019$	$R = 0.79, P < 0.001$	$R = -0.53, P = 0.028$	*				
Maternal worries		$R = -0.47, P = 0.035$		$R = 0.55, P = 0.019$	*			
Paternal worries			$R = -0.51, P = 0.025$	$R = 0.70, P = 0.002$	$R = 0.67, P = 0.002$	*		
Maternal distress		$R = -0.72, P < 0.001$	$R = -0.57, P = 0.012$	$R = 0.49, P = 0.046$	$R = 0.75, P < 0.001$	$R = 0.62, P = 0.002$	*	
Paternal distress		$R = -0.69, P = 0.001$	$R = -0.71, P = 0.001$	$R = 0.54, P = 0.025$	$R = 0.59, P = 0.008$	$R = 0.63, P = 0.004$	$R = 0.85, P < 0.001$	*

Due to improvements in treatment facilities since the early 1970s, haemophilia has become a controllable disorder that no longer automatically results in severe impairments of the joints and early death (e.g. [5]). Therefore, one may expect to find lower levels of worries and illness-related distress in patients and in parents of patients with haemophilia, compared to a chronic illness that is still associated with high mortality risks. Indeed, we found relatively high-QoL scores in both patients and parents. Moreover, compared to patients with Cystic fibrosis [17], the patients in our study are less worried about the transition.

Theoretically, impediments to the transition process may arise from both patients and their parents (e.g. [6,8,9,11–13]). Our findings indicate that pre- and posttransition patients do not differ in self-reported QoL or in worries about the transition. Attanasio *et al.* [21] also found no effects of the transition on QoL in patients with childhood onset growth hormone deficiency. However, in patients with cystic fibrosis, Boyle *et al.* [17] found significantly lower worries in posttransition patients than in pretransition patients. This discrepancy may possibly be explained by the larger sample size in the study by Boyle *et al.* [17]. Alternative explanations may be cultural differences between the Netherlands and the USA or illness-related differences (e.g. differences in medical prognosis; see above). While no differences were found between pre- and posttransition patients, differences in QoL and illness-related distress were found between parents of pre- and posttransition patients. Fathers of posttransition patients reported their sons' QoL to be lower than fathers of pretransition patients. We also observed a tendency in the same direction for mothers. It is possible that parents are more sensitive to changes in their child's QoL than the child is himself. However, alternative explanations exist. For instance, the transition from paediatric to adult health care services may have negative effects on the parent's perceived control with respect to their son's medical condition. In chronically ill patients, it has been found that perceived control over the medical condition is associated with better QoL (e.g. [22,23]). One can argue that, in parents of chronically ill children, a decrease in their perceived control over their child's medical condition negatively affects their perception of the child's medical condition and QoL status. In line with this explanation, we found that fathers of posttransition patients also reported higher illness-related distress compared to fathers of pretransition patients. Moreover, our findings indicate that parents use different signals to rate their son's

QoL than their son does himself; parental estimations of their son's QoL were associated with their son's self-rated QoL and with their own and their partner's worries and illness-related distress. The patient's self-rated QoL was found to be associated only with his parents' QoL ratings.

Our findings indicate that mothers tend to be more worried about the transition than fathers and patients, in particular with respect to medical issues around the transition. Saviolo-Negrin *et al.* [24] found that mothers are more depressed and anxious about their haemophilic child's disease than fathers. Bottos *et al.* [25] found that fathers and mothers respond differently to a psychological support and counselling programme for parents with haemophilic children. Based on these findings, one could argue that programmes designed to facilitate parents in the transition process from paediatric to adult health care services require a gender-specific approach.

The present study has some limitations that need to be discussed. First, our sample size is small and the study was cross-sectional in design. Therefore, our findings clearly need to be replicated in longitudinal studies. Secondly, in general, the participating patients' health-related QoL, as reported in self-reports and the parents' reports, can be considered high. Also, patients and parents reported relatively few worries about the transition. Furthermore, parental illness-related distress can be considered as low to moderate. This may indicate that the population under study is doing fairly well. We cannot exclude the possibility that our findings will turn out to be different under less favourable health care circumstances. Another limitation is the number of drop outs. Of course, one cannot exclude the possibility that reasons for drop out may have a relationship with the effects of the transition. However, we feel that the main argument for drop out (lack of time) fits within the general finding that for the present sample the transition from paediatric to adult health care services does not have a strong impact on patients and their parents. Finally, we did not investigate the patients' medical condition. Health-related QoL is associated with the patient's medical condition (e.g. [26]). However, we cannot exclude the possibility that the transition affects the medical condition in ways that are not noticed by questionnaires that assess QoL.

Our findings indicate that the transition from paediatric services to adult health care facilities appears to have stronger impact on parents of patients with haemophilia than on the patients themselves. Empowerment programmes aimed at parents can help to ease their worries about the transfer of their child

and to avoid increases in illness-related distress. Gender differences in how parents respond to their child's illness need to be taken into consideration in the development of these programmes.

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## Disclosures

The authors stated that they had no interests which might be perceived as posing a conflict or bias.

## References

- 1 Mannucci PM, Tuddenham EGD. The hemophiliac – from royal genes to gene therapy. *N Engl J Med* 1991; **344**: 1733–79.
- 2 Bolton-Maggs P, Pasi JK. Haemophilias A and B. *Lancet* 1993; **361**: 1801–9.
- 3 Blum RW. Transition to adult health care: setting the stage. *J Adolesc Health* 1995; **17**: 3–5.
- 4 Suris JC. Global trends of young people with chronic and disabling conditions. *J Adolesc Health* 1995; **17**: 17–22.
- 5 Tiemstra AHM, Smit C, van der Ploeg HM, Briët E, Roosendaal FR. Two decades of haemophilia treatment in the Netherlands, 1972–1992. *Haemophilia* 1995; **1**: 165–71.
- 6 Flemming E, Carter B, Gillibrand W. The transition of adolescents with diabetes from the children's health care service into the adult health care service: a review of the literature. *J Clin Nurs* 1992; **11**: 560–7.
- 7 Binks JA, Barden W, Burke TA, Young NL. What do we really know about the transition to adult-centered health care? A focus on cerebral palsy and spina bifida. *Arch Phys Med Rehabil* 1997; **88**: 1064–73.
- 8 Rosen DS. Between two worlds: bridging the cultures of child health and adult medicine. *J Adolesc Health* 1995; **17**: 10–6.
- 9 Viner R. Transition from paediatric to adult care. Bridging the gap or passing the buck?. *Arch Dis Child* 1999; **81**: 271–5.
- 10 Hobbie WL, Ogle S. Transitional care for young adult survivors of childhood cancer. *Semin Oncol Nurs* 1991; **17**: 268–73.
- 11 Rosen DS. Transition of young people with respiratory diseases to adult health care. *Pediatr Respir Rev* 1994; **5**: 124–31.
- 12 McDonagh JE. Growing up and moving on: transition from pediatric to adult care. *Pediatr Transplant* 1995; **9**: 364–72.

- 13 Scal P, Ireland M. Addressing transition to adult health care for adolescents with special health care needs. *Pediatrics* 1995; **115**: 1607–12.
- 14 Calahan ST, Feinstein Winitzer R, Keenan P. Transition from pediatric to adult oriented health care: a challenge for patients with chronic disease. *Curr Opin Pediatr* 1991; **13**: 310–6.
- 15 Rosen DS, Blum RW, Britto M, Sawyer SM, Siegel DM. Transition to adult health care for adolescents and young adults with chronic conditions. *J Adolesc Health* 1993; **33**: 309–11.
- 16 Bennet DL, Towns SJ, Steinbeck KS. Smoothing the transition to adult care. *MJA* 1995; **182**: 373–4.
- 17 Boyle MP, Faqruckhi Z, Nosky ML. Strategies for improving transition to adult cystic fibrosis care, based on patient and parent views. *Pediatr Pulmonol* 1991; **32**: 428–36.
- 18 Flood E, Szende A, Rentz A *et al.* Health-related quality of life in adults with hemophilia: the patient perspective. *J Thromb Haemost* 2003; **1** ( Suppl. 1): P0657.
- 19 van Dongen-Melman JEW. On Surviving Childhood Cancer. Late Psychological Consequences for Patients, Parents, and Siblings. PhD Thesis, Erasmus University Rotterdam, Rotterdam, 1995.
- 20 Cohen J, Cohen P. *Applied Multiple Regression/Correlation Analysis for the Behavioral Sciences*. Hillsdale, NJ: Erlbaum, 1983.
- 21 Attanasio AF, Shavrikova EP, Blum WF, Shalet SM. Quality of life in childhood onset growth hormone-deficient patients in the transition phase from childhood to adulthood. *J Clin Endocrinol Metab* 1995; **90**: 4525–9.
- 22 Affleck G, Tenne H, Pfeifer C, Fifield J. Appraisals of control and predictability in adapting to a chronic disease. *J Pers Soc Psychol* 1987; **53**: 273–9.
- 23 Culos-Reed SN, Brawley LR. Fibromyalgia, physical activity, and daily functioning: the importance of efficacy and health related quality of life. *Arthritis Care Res* 1999; **13**: 343–51.
- 24 Saviolo-Negrin N, Cristante F, Zanon E, Canclini M, Stocco D, Girolami A. Psychological aspects and coping of parents with a haemophilic child: a quantitative approach. *Haemophilia* 1999; **5**: 63–8.
- 25 Bottos ASM, Zanon E, Sartori MT, Girolami A. Psychological aspects and coping styles of parents with haemophilic child undergoing a programme of counselling and psychological support. *Haemophilia* 1997; **13**: 305–10.
- 26 Mackensen S von, Bullinger M, The HAEMO-QOL Group. Development and testing of an instrument to assess the quality of life of children with haemophilia in Europe (Haemo-QoL). *Haemophilia* 1994; **10**: 17–25.